

## A 37-year-old Man with a Painless Growing Mass of the Thorax

Boris Michael Holzapfel MD, Christoph Schaeffeler MD,  
Ingo Jörg Banke MD, Simone Waldt MD

Received: 19 March 2009 / Accepted: 10 August 2009 / Published online: 28 August 2009  
© The Association of Bone and Joint Surgeons® 2009

### History and Physical Examination

A 37-year-old Indian man presented with painless swelling of the right dorsolateral thorax. The lesion initially was noticed approximately 12 months earlier. Since then, it had grown continuously in size. An open biopsy at another institution obtained one month after onset provided non-diagnostic material. On presentation to us, the patient denied constitutional symptoms, such as fever, night sweats, fatigue, or recent weight loss. The patient's physical status was good and his medical and family history were noncontributory. He had no history of other masses. The patient was working as an electrical engineer and had moved to Germany 10 months before consultation.

Physical examination revealed a well-defined tender mass approximately 10 × 10 cm, which could be displaced toward the musculature. There were no local inflammatory

signs or lymphadenopathy. The 2 cm longitudinal scar from the previous biopsy was well healed without hypertrophy or redness.

Imaging studies were performed and included initially chest radiography and MRI (Figs. 1–3). The radiograph of the chest was unremarkable.

Based on the history, physical examination, and imaging studies, what is the differential diagnosis?

### Imaging Interpretation

MRI revealed a 5- × 3.8- × 5.2-cm oval mass located in the serratus anterior muscle. The mass was fluid-equivalent with intermediate signal on T1-weighted images (Fig. 1) and high signal on T2-weighted images (Fig. 2). There was a small amount of surrounding edema. In the larger cystic formation, a distinct lesion could be detected, showing a liquid-equivalent signal and with a size of 1.4 × 1.2 cm; in this small lesion, a solid hypointense area was seen in all sequences. The multilayered margin of the outer cyst intensely took up contrast medium (Fig. 3).

An ultrasound examination revealed a hypoechoic outer lesion. In the inner lesion, we observed hyperechoic areas corresponding with the hypointense structures found on MRI and interpreted these as calcifications. Doppler ultrasound examination revealed no signs of vascularity.

The patient underwent complete staging using total-body MRI to exclude a systemic disease. No other lesions were found.

Laboratory tests showed no abnormal findings, but enzyme-linked immunoblot assay was positive for cysticercosis antibodies.

---

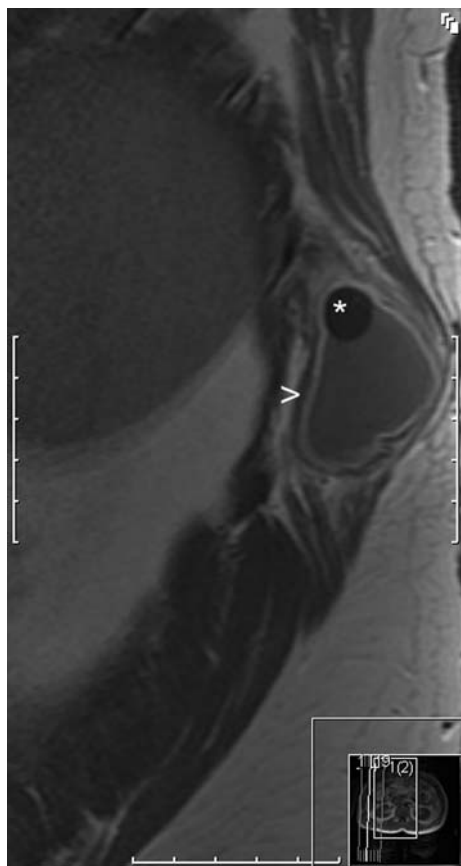
Each author certifies that he or she has no commercial associations (eg, consultancies, stock ownership, equity interest, patent/licensing arrangements, etc) that might pose a conflict of interest in connection with the submitted article.

Each author certifies that his or her institution approved or waived approval for the reporting of this case report and that all investigations were conducted in conformity with ethical principles of research.

---

B. M. Holzapfel (✉), I. J. Banke  
Department of Orthopaedic Surgery, Klinikum Rechts der Isar,  
Technical University Munich, Ismaninger Str 22, D-81675  
Munich, Germany  
e-mail: Boris.Holzapfel@gmx.de

C. Schaeffeler, S. Waldt  
Institute of Radiology, Klinikum Rechts der Isar, Technical  
University Munich, Munich, Germany



**Fig. 1** A sagittal T1-weighted turbo spin-echo MR image shows a cystic intramuscular lesion with intermediate signal intensity (>) and inside of it a smaller cyst with low signal intensity (\*).

### Differential Diagnosis

- Myxoid neoplasm
- Solitary parasitic cyst (hydatid disease, cysticercosis)
- Metastatic disease
- Abscess

As the lesion was unifocal, a wide surgical resection was chosen. The skin was incised carefully and a cystic mass identified in the muscle. During the dissection, the mass was not violated and the lesion was totally excised with wide margins (Fig. 4). The lamellar cyst wall could be completely preserved. Histologic evaluation was obtained (Fig. 5).

Based on the clinical history, physical examination, radiographic images, and histologic examination, what is the diagnosis and how should the lesion be treated?

### Histopathologic Interpretation

Macroscopically, a cystic mass with myxoid viscous yellowish material was seen (Fig. 4). Microbiologic analysis

and Gram stain revealed leukocytes but no organisms. There was no growth from the culture. After lavation, a smaller cyst in the larger cyst was obvious. In the cavity of the smaller cystic lesion, there was a yellow papillary growth infiltrating the wall of the cyst.

Microscopic analysis revealed a cysticercus with an invaginated white scolex (Fig. 5A) in the smaller cyst. An inflammatory mass with lymphofollicular infiltrate surrounded the cysticercus. The cystic wall was interspersed predominantly with numerous macrophages and necrotic areas.

The typical four suckers and the double row of hooks could be verified (Fig. 5B–C).

### Diagnosis

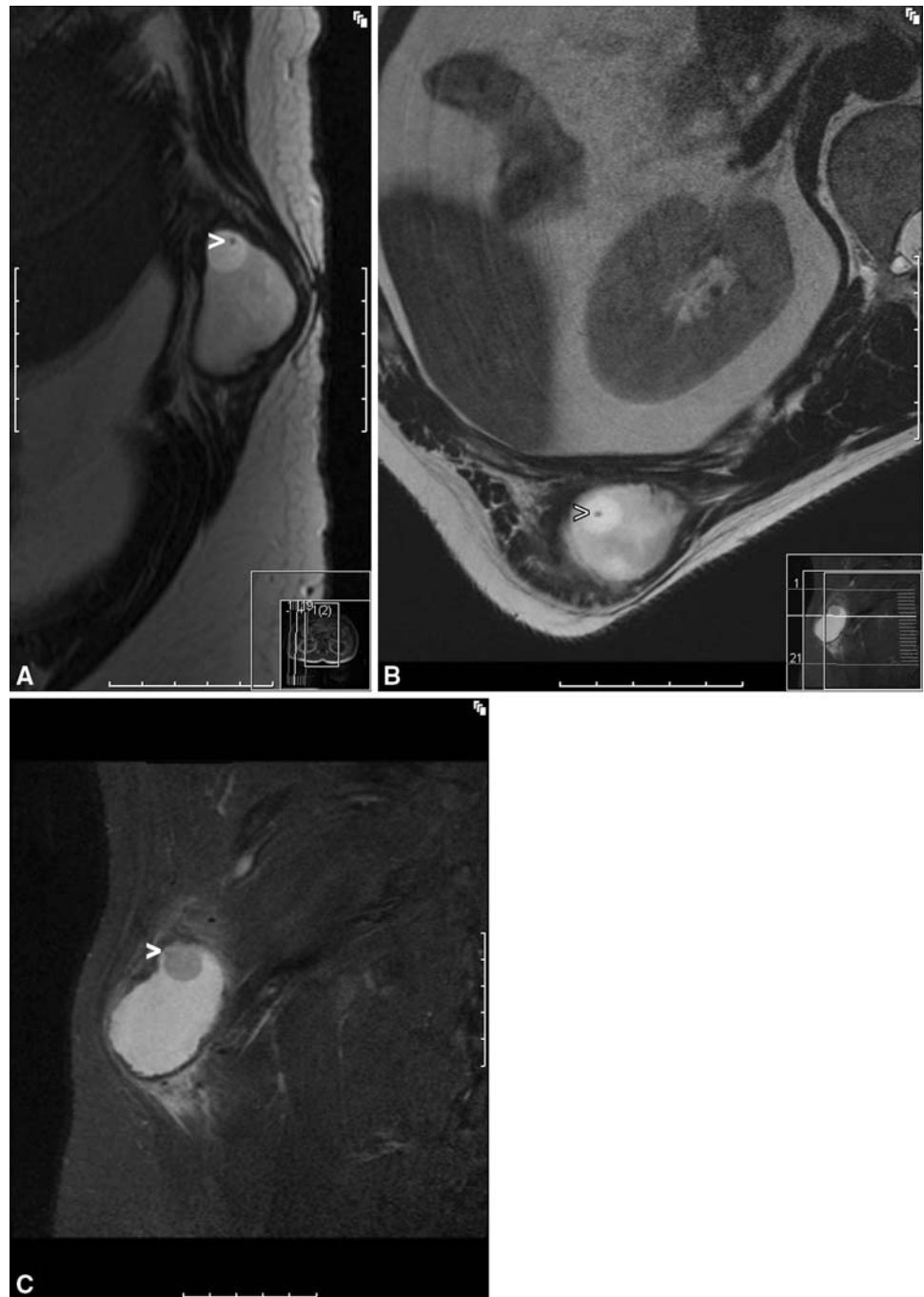
Solitary cysticercus of the thorax musculature presenting as a tumor.

### Discussion and Treatment

The differential diagnosis based on clinical and radiographic data included a myxoid neoplasm, parasitic cyst, abscess, and metastatic disease. Although metastatic disease or myxoid neoplasm was a consideration, MR images of the soft tissue mass were more consistent with a solitary parasitic cyst or an abscess. These typical features were fluid-equivalent signal and peripheral contrast medium enhancement of the capsule. A solid central lesion could be seen, which suggested the presence of a scolex. However, imaging studies could not clearly distinguish between an abscess and a parasitic cyst. Nevertheless, the possibility of an infection was considered low because there were no systemic or local inflammatory signs. The patient's state of health was good and did not indicate a consuming disease such as metastatic cancer.

Myxoid tumors of soft tissue encompass a heterogeneous group of lesions characterized by a marked abundance of extracellular mucoid matrix. These tumors show substantial variability in their biologic behavior, thus including lesions that are entirely harmless, tumors with a tendency to locally recur but do not metastasize, and malignant tumors. Because of the considerable degree of overlap clinically and morphologically between the various tumor types in this group, potential diagnostic problems often are generated. In this case, the diagnosis of a myxoid tumor could be excluded only with certainty by performing a biopsy. But, with the possibility of a parasitic cyst in mind, we decided not to perform an incision biopsy to prevent systemic dissemination of the parasite.

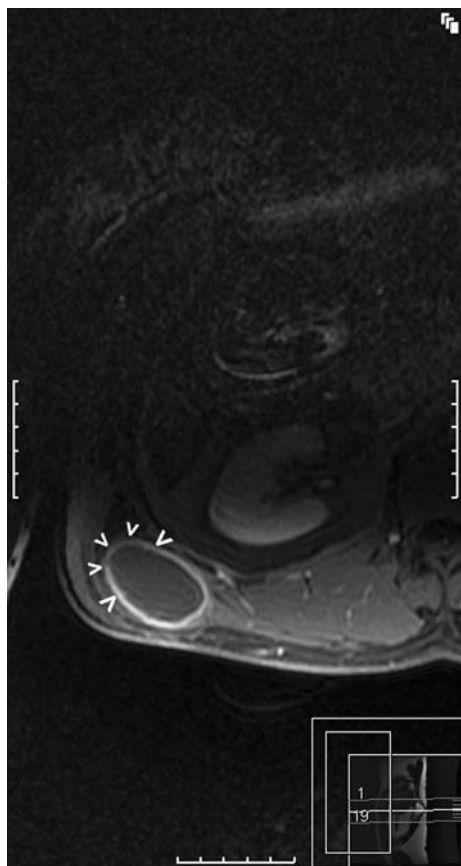
**Fig. 2A–C** On (A) sagittal and (B) axial T2-weighted turbo spin-echo images and (C) a coronal STIR sequence, a small dot (>) can be seen, which is consistent with a scolex.



The lesion was nonspecific, but a parasitic cyst was favored. It could be confirmed by positive enzyme-linked immunoblot assay, but it did not definitely indicate the presence of living parasites. Antibodies can be seen to persist for a long time in immune individuals after the death of the parasites. Finally, histologic evaluation of the tissue obtained after surgery established the diagnosis of solitary cysticercosis of the serratus anterior muscle encapsulated by a purulent cyst.

*Taenia solium* infection and the resulting symptoms are endemic in less developed countries with poor hygiene

standards where pigs are raised as a food source. Cysticercosis is common in Mexico, Asia, Central and South America, India, and parts of Africa [18]. *T. solium*, also called the pork tapeworm, is a cyclophyllid cestode in the family Taeniidae. It has a complex two-host life cycle. Cysticercosis in pigs as regular intermediate hosts is caused by ingestion of food contaminated with eggs containing feces of human tapeworm carriers. When ingested, eggs develop into larvae in the intestine of the animal. These larvae migrate through the intestinal wall into the blood circulation and localize in various tissues and organs by



**Fig. 3** An axial gadolinium-enhanced T1-weighted spin-echo MR image shows the enhancing margin ( $>$ ) of the outer cyst.

forming multiple cysts. When these encysted larvae die, they can induce a granulomatous inflammatory response, which can become symptomatic.

Humans are the only definitive host and harbor the adult tapeworm. After ingestion of undercooked pork infected with cysticerci, these develop into adult worms in the human host. Attaching to the intestinal mucosa, the parasites release proglottids containing eggs or oncospheres. These then can enter the intermediate host, thus completing the parasite's life cycle [6]. Apart from being the definitive host, humans can act as the intermediate host by consumption of food or water contaminated with eggs or proglottids. Autoinfection by regurgitation of proglottids into the stomach has been proposed but not proven [12].

Neurocysticercosis is the most prevalent infection of the central nervous system worldwide and is the greatest cause of acquired epilepsy worldwide [19]. Less frequently, other organs are involved, including muscles, subcutaneous soft tissues, the eyes, the oral cavity, the heart, and the lungs [9, 11, 13, 20]. Mostly multilocular muscular disease with subsequent pseudohypertrophy caused by multiple cysticercosis is reported.



**Fig. 4** A photograph shows the excised tumor with wide margins and a test tube filled with thick yellowish liquid obtained by puncture of the intact mass.

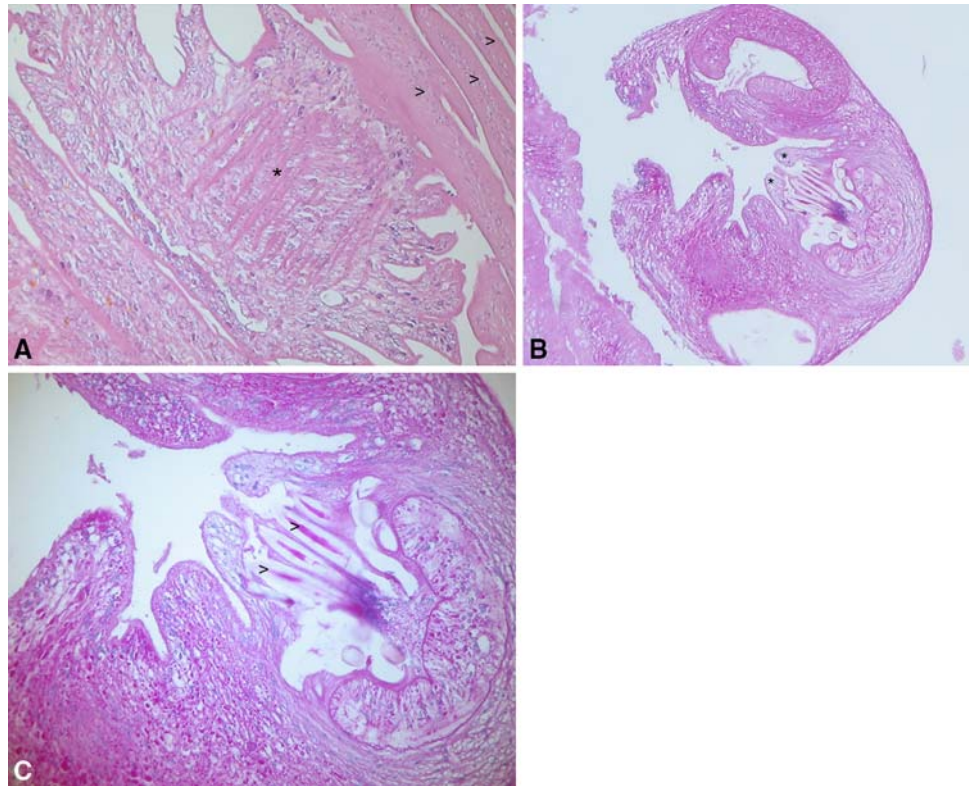
In contrast, isolated muscular involvement by only one pseudocyst is rare. Only 14 cases have been described to date [1–3, 5, 7, 8, 10, 14, 22], and none of these occurred in Central Europe. Because of the unspecific symptoms, isolated soft tissue cysticercosis is very difficult to diagnose. The encysted larvae may cause no symptoms for a long time. Usually the disease is confused with other benign swellings. Normally the cysts vary in size from a few millimeters to 1 to 2 cm [6]. However, in our patient, the cyst reached several centimeters in diameter.

To establish a diagnosis, a history of residence in an endemic country can be helpful. Our patient moved to Germany from India 10 months before consultation. But, as an engineer, he had no contact with animals used for food. He reported eating no raw or uncooked meat.

Routine laboratory tests rarely contribute, which can be documented in our patient. Until recently, the attempt at serologic diagnosis of cysticercosis using enzyme-linked immunosorbent assay has been less than satisfactory. The enzyme-linked immunoblot assay performs much better and is reported to have a sensitivity of 98% and specificity of 100% [15, 16]. However, in patients with only a few degenerating cysts or calcified lesions, its sensitivity is much lower [3, 16].

MRI seems to be the preferred diagnostic procedure although CT is more sensitive to detect calcifications. MRI is the most accurate technique to assess the degree of infection, the location, and the evolutionary stage of the cysticercus. Usually it shows characteristic structures of the cyst. A round area approximately 1 cm in diameter with a low signal on T1-weighted images and a high signal on T2-weighted images can be found. In this area, a small dot containing the scolex can be seen. However, images often differ from this owing to the life cycle stages of the parasite

**Fig. 5A–C** (A) A high-power photomicrograph (Stain, hematoxylin and eosin, original magnification,  $\times 40$ ) reveals parts of the cyst wall ( $>$ ) and the region of the inverted scolex (\*). In (B) a low-power view (Stain, periodic acid-Schiff; original magnification,  $\times 16$ ) and (C) a high-power view (Stain, periodic acid-Schiff; original magnification,  $\times 40$ ), the hooklets ( $>$ ) and the rostellum (\*) of the scolex are clearly visible.



and the response activity by the host [17]. In response to an inflammatory process, the cysticercus can be embedded in a pseudocyst, as it was in our patient. Peripheral contrast medium enhancement in the adjacent tissue indicated this.

Many reports of solitary muscular and soft tissue cysticercosis lack adequate staging for other cysts. In most cases, where only a solitary cyst was described, only plain radiography and CT scans of the cranium were applied to exclude other infestation sites [1–3, 5, 7, 8, 10, 14, 22]. We consider this insufficient and recommend total-body MRI.

For symptomatic solitary cysts outside the central nervous system, many doctors advocate surgical resection. Although encysted larvae do not always result in clinical symptoms, systemic therapy usually is advocated in the presence of multilocular cysts or if the number of cysts makes surgical therapy unfeasible or neurocysticercosis is verifiable. In these cases, systemic therapy, for example, with antiparasitic drugs, such as praziquantel and albendazole, should be used [4, 6, 14]. Praziquantel (50 mg/kg per day for approximately 3 weeks) is reportedly effective and is considered the preferred drug for treatment of cysticercosis, although some studies indicate albendazole (10–15 mg/kg per day for approximately 2 weeks) shows better response rates [6, 21].

In the case of a muscular tumor of unknown etiology, particularly in a patient from an endemic area, cysticercosis and other parasitic cysts should be considered as a possible cause. Because of increasing globalization and

labor mobility, orthopaedic surgeons in Western countries also should be aware of parasitic diseases.

When features of a solitary cysticercus are present on MRI, enzyme-linked immunoblot assay should be performed, to help find the correct diagnosis. In case of a solitary cysticercus, surgical resection is the preferred therapy. Total-body MRI to exclude additional infestation sites is highly recommended for choosing the best therapeutic regime. In the case of a multilocular pattern, systemic therapy with antiparasitic drugs is indicated complementary to surgical therapy.

As our patient was free of other relevant symptoms and a systemic or multilocular appearance of the disease was excluded, no additional treatment was necessary after an uneventful postoperative period. The patient was informed about the nature of the disease, regular periodic checkups were suggested, and he was returned to outpatient care. At last followup 1 year postoperatively, he had no local or systemic complaints.

**Acknowledgments** We thank Hakan Pilge and Hans Gollwitzer for assistance with preparation of this manuscript.

## References

1. Abdelwahab IF, Klein MJ, Hermann G, Abdul-Quader M. Solitary cysticercosis of the biceps brachii in a vegetarian: a rare or unusual pseudotumor. *Skeletal Radiol.* 2003;32:424–428.

2. Anderson GA, Chandi SM. Cysticercosis of the flexor digitorum profundus muscle producing flexion deformity of the fingers. *J Hand Surg Br.* 1993;18:360–362.
3. Brown ST, Brown AE, Filippa DA, Coit D, Armstrong D. Extraneural cysticercosis presenting as a tumor in a seronegative patient. *Clin Infect Dis.* 1992;14:53–55.
4. Brown WJ, Voge M. Cysticercosis: a modern day plague. *Pediatr Clin North Am.* 1985;32:953–969.
5. Ergen FB, Turkbey B, Kerimoglu U, Karaman K, Yorganc K, Saglam A. Solitary cysticercosis in the intermuscular area of the thigh: a rare and unusual pseudotumor with characteristic imaging findings. *J Comput Assist Tomogr.* 2005;29:260–263.
6. Garcia HH, Gonzalez AE, Evans CA, Gilman RH; Cysticercosis Working Group in Peru. *Taenia solium* cysticercosis. *Lancet.* 2003;362:547–556.
7. Jankharia BG, Chavhan GB, Krishnan P, Jankharia B. MRI and ultrasound in solitary muscular and soft tissue cysticercosis. *Skeletal Radiol.* 2005;34:722–726.
8. Kazanjian PH, Mattia AR. Case records of the Massachusetts General Hospital. Weekly clinicopathological exercises. Case 26–1994. A 20-year-old Philippine woman with a soft-tissue mass in the forearm. *N Engl J Med.* 1994;330:1887–1893.
9. Kraft R. Cysticercosis: an emerging parasitic disease. *Am Fam Physician.* 2007;76:91–96.
10. Kung IT, Lee D, Yu HC. Soft tissue cysticercosis: diagnosis by fine-needle aspiration. *Am J Clin Pathol.* 1989;92:834–835.
11. Malik SR, Gupta AK, Choudhry S. Ocular cysticercosis. *Am J Ophthalmol.* 1968;66:1168–1171.
12. Martinez MJ, de Aluja AS, Gemmell M. Failure to incriminate domestic flies (Diptera: Muscidae) as mechanical vectors of *Taenia* eggs (Cyclophyllidae: Taeniidae) in rural Mexico. *J Med Entomol.* 2000;37:489–491.
13. Mehta DS, Malik GB, Dar J. Intramedullary cysticercosis. *Neurol India.* 1971;19:92–94.
14. Ogilvie CM, Kasten P, Rovinsky D, Workman KL, Johnston JO. Cysticercosis of the triceps—an unusual pseudotumor: case report and review. *Clin Orthop Relat Res.* 2001;382:217–221.
15. Proano-Narvaez JV, Meza-Lucas A, Mata-Ruiz O, Garcia-Jeronimo RC, Correa D. Laboratory diagnosis of human neurocysticercosis: double-blind comparison of enzyme-linked immunosorbent assay and electroimmunotransfer blot assay. *J Clin Microbiol.* 2002;40:2115–2118.
16. Rosas N, Sotelo J, Nieto D. ELISA in the diagnosis of neurocysticercosis. *Arch Neurol.* 1986;43:353–356.
17. Salgado P, Rojas R, Sotelo J. Cysticercosis: clinical classification based on imaging studies. *Arch Intern Med.* 1997;157:1991–1997.
18. Sawhney IM, Singh G, Lekhra OP, Mathuriya SN, Parihar PS, Prabhakar S. Uncommon presentations of neurocysticercosis. *J Neurol Sci.* 1998;154:94–100.
19. Shandera WX, Kass JS. Neurocysticercosis: current knowledge and advances. *Curr Neurol Neurosci Rep.* 2006;6:453–459.
20. Singh I, Phogat AC, Chohan BS, Malik KP. Conjunctival cysticercosis. *J Indian Med Assoc.* 1978;70:136–137.
21. Sotelo J, Del Brutto OH, Penagos P, Escobedo F, Torres B, Rodriguez-Carbajal J, Rubio-Donnadieu F. Comparison of therapeutic regimen of anticysticercal drugs for parenchymal brain cysticercosis. *J Neurol.* 1990;237:69–72.
22. Yue XH. Fine needle aspiration biopsy diagnosis of cysticercosis: a case report. *Acta Cytol.* 1994;38:90–92.